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CLINICAL REPORT

Surgical treatment of the symptomatic os trigonum in children

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Accepted: 8 October 2008

KEYWORDS

Os trigonum;
Children;
Posterior ankle
impingement;
Talus ossicle

Summary Symptomatic os trigonum is a rare condition, well described in adults, that causes chronic ankle pain. To date there are no reported cases of successfully managed symptomatic os trigonum in the children population. We retrospectively reviewed four paediatric patients (11–17 years of age) successfully operated for a symptomatic os trigonum using an open excision through a posteromedial approach. One case was bilateral. Postoperative pain relief was obtained in all cases. All of the patients were able to return to unrestricted physical activities after three months. The average follow-up was 12 months. Symptomatic os trigonum may be held responsible for chronic ankle pain in children and adolescents as well as in adults. The surgical treatment is effective in children.

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Introduction

The os trigonum is an accessory bone with inconstant presence at the posterolateral side of the talus. It appears between the ages of eight and 11 years as a nucleus of secondary ossification that generally fuses in less than one year. It is classically triangular with anterior, inferior, and posterior sides. It can also be rounded or oval. It is 10 mm long and 10 mm high but can vary if it is in several parts [1]. Laterally, it receives fibers from the talofibular ligament and is in contact medially with the flexor hallucis longus sheath. It can persist either because of the absence of fusion of

the ossification nucleus [2] or a fracture of the trigonum apophysis, which later is not able to consolidate.

Os trigonum syndrome usually involves young and athletic adults. We have not found any publications in the literature on this syndrome specifically in children. However, this disease does exist in children and adolescents. The clinical picture does not differ from the symptoms in adults. Although management in adults has remained controversial, we have had a decisively surgical attitude, after the failure of a well-conducted medical treatment. This tactic has been entirely satisfactory.

Observation

We report four cases of adolescent patients, three females and one male, who consulted for chronic handicapping pain

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Table 1 Summary of the patients.

Patients	Age	Sports activities	Reason for consultation	Symptoms	Imaging studies	Preoperative treatments	Surgical treatments	Results
1	11 years	Dancing, swimming	Pain, both feet: – Duration: 1 year – No injury	Pain at plantar flexion bilaterally	X-rays MRI Scintigraphy	Rest 3 months: no effect Infiltration of corticosteroids: no pain for 3 weeks	Surgical excision by medial approach bilaterally	Pain relief resumption of sports activities at M3
2	15 years	High-level fencing	Pain, right foot: – Duration: 1 year – Initial injury: ankle sprain	Internal retromalleolar pain increased at plantar flexion	X-rays MRI	Cessation of sports activity NSAID	Surgical excision by medial approach	Pain relief, resumption of sports activities at M3
3	14 years	Running	Pain, posterior side of left ankle Duration: 10 months	Increased when climbing and descending stairs	X-rays CT MRI Scintigraphy	Cessation of sports activity NSAID	Surgical excision by medial approach	Pain relief resumption of sports activities at M3
4	17 years	Football	Left talocalcaneal pain Duration: 1 year	Talagia increased with plantar flexion Pain on flexion of hallux against resistance	Echography Radiography MRI	Cessation of sports activity NSAID	Surgical excision by medial approach	Pain relief, resumption of sports activities at M3

in the ankle and foot. The radiological workup demonstrated the presence of os trigonum responsible for the symptoms. Surgical treatment eliminated the pain, with a mean follow-up of 1 year (± 2 months). The data for these four patients are provided in Table 1.

Operative technique

The patient is placed in the dorsal decubitus position, with the lower limb placed in flexion and external rotation of the hip for easy exposure of the internal retromalleolar area. It may be useful to identify the os trigonum using the image intensifier.

The incision is posteromedial in a retromalleolar curve, 4 cm long. The quadratus muscle of the toes is identified first, then the posterior tibial neurovascular bundle, and finally the flexor hallucis longus, which are put on a tie. The traction on the flexor hallucis longus exposes the os trigonum. Using a small elevator defines its edges. It is excised easily with a scalpel. We begin by detaching the posterior talofibular ligament insertion to prevent the os trigonum from moving into the external retromalleolar region when this ligament is retracted. After surgery, we allow weight-bearing as soon as pain is relieved (on average the third postoperative day).

Discussion

Os trigonum is a rare pathology since most of the series only report a small number of adult patients and a review of the literature only found a few cases in adolescents [3–7]. The mean patient age in the longest series reported (43 cases) was 27 years and the youngest patient was 16 years old [7]. Our series is the first pediatric series.

The physiopathology of os trigonum syndrome calls into question an exaggerated plantar flexion that is either acute – this is the equivalent of a fracture of the posterior process of an overly long talus – or chronic [8]. Sports requiring forced plantar flexion such as dancing (on points) and football (shooting) are sports that classically cause problems [9–11]. Three of our four patients took part in sports with forced plantar flexion.

The clinical presentation of the four pediatric cases reported herein did not seem to differ from the presentation found in the adult population. The time to diagnosis is often extended by several weeks or months. The disorder usually involves relatively unsystematic pain in the posterior region of the heel, with the seat of pain in the external retromalleolar region, or sometimes internal. The pain is reproduced in maximal plantar flexion. Resisted flexion of the hallucis can also trigger pain (particularly if there is associated tenosynovitis of the flexor hallucis).

In the general population, the presence of os trigonum varies from 2 to 50% [12]. Pain in the hindfoot with presence of an os trigonum on the lateral X-ray of the ankle does not suffice to establish the diagnosis of os trigonum syndrome. The problem is confirming the os trigonum as the cause of pain [7].



Figure 1 IRM patient n° 1.

This problem is even more difficult to treat in the pediatric population than in adults because of the physiological presence of an ossification nucleus. MRI, scintigraphy, and the infiltration test of local anesthetics are three means that can demonstrate os trigonum involvement [7,13,14]. The infiltration test of the os trigonum using lidocaine is the preferred test done with image intensifier guidance [13]. Hyperfixation on scintigraphy is a good argument for os trigonum involvement, despite authentic false-negative and false-positive results [7,14].

MRI is superior to CT in this indication. According to Tamburini et al. [15], MRI provides specific images of tissue damage in the os trigonum zone, such as increased size of the synchondrosis between the talus and the os trigonum, medullary edema of the os trigonum, and less specific signs such as inflammation of the posterior tibiotalar capsule and tenosynovitis of the flexor hallucis longus (Fig. 1). Moreover, MRI can explore the os trigonum and the adjacent soft tissues; it can provide a differential diagnosis with posterior ankle impingement syndrome [7]. We found signal anomalies within the os trigonum or in the surrounding area in 100% of the cases presented.

After taking plain X-rays demonstrating the os trigonum, we recommend bone scintigraphy (Fig. 2A), which can be used to orient CT exploration (Fig. 2B) or MRI, particularly in children in whom taking the patient's history and the clinical examination are more difficult. This second series of exams confirms the os trigonum as the cause of pain.

We carried out lidocaine infiltration guided by the image amplifier only once in this series (in the case with the longest history). The problems performing this act in pediatric patients without general anesthesia and the increasingly accessible MRI motivated our abandoning this invasive exploration technique.

Managing os trigonum syndrome begins by discontinuing sports, with foot and leg support associated with nonsteroidal anti-inflammatory drugs [5,9–11]. Immediate surgery is not recommended.

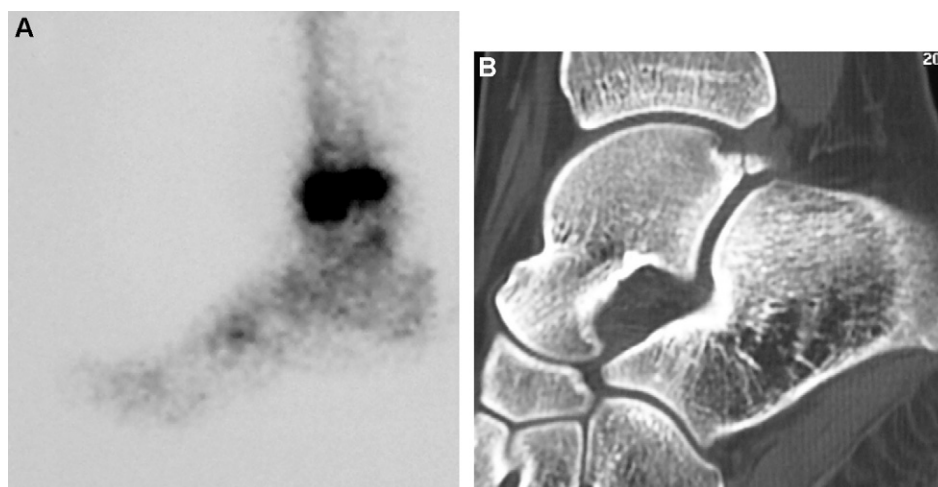


Figure 2 (A and B) patient n° 3.

Some authors recommend local anesthetic and cortisone infiltration [9,10]. If conservative treatment fails, the os trigonum must be resected surgically. The orthopedic option should not excessively delay surgery: it has been demonstrated that waiting longer than two years before surgery is a risk factor for a less satisfactory functional result [7].

The os trigonum can be resected in conventional surgery or in arthroscopy. The posterolateral surgical approach seems to be the most frequently used [7,9,13]. It can be complicated by neuromas or dysesthesias in the short saphenous nerve area. However, systematically identifying this nerve reduces the incidence of these complications [16]. Like us, certain authors [10,17] nevertheless recommend the posteromedial surgical approach. All the structures that must be respected during surgical resection of the os trigonum (posterior tibial neurovascular pedicle, the common flexor of the toes, flexor digitorum longus, flexor hallucis longus) are easily visualized from this approach, which does little functional damage and whose scar is very small and faint (4cm). We believe this approach to be safer than the posterolateral approach that is classically described in the literature. We have observed no nerve complications (in either the posterior tibial area or the sural nerve area). It should be noted that this anatomic region is easily explored by surgeons who are experienced in operating on infants with club foot.

Arthroscopic resection is technically demanding and should only be undertaken by experienced arthroscopist surgeons [17–19]. The result is equivalent at the medium and long term, but arthroscopic techniques heal more quickly and allow the patient to return to sports activity more rapidly (3 months instead of 6 months) [20,21].

Several arthroscopic techniques have been described for the various approaches used. Combining both the posterolateral and posteromedial approaches [22], thus superimposing two posterolateral approaches [19], allows resecting the os trigonum without penetrating the talofibular joint.

In our series, the time to resuming a sports activity was comparable to the times observed after conventional surgery in adult series. In view of the endoscopic results, we do not exclude envisioning these techniques for future cases.

Conclusion

The os trigonum is most often asymptomatic. When symptoms appear in children, they are similar to those reported in adults. The definitive diagnosis should be based on complementary scintigraphic and MRI exams. Treatment can be medical at first, but should not delay surgical management if it is not successful.

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